

# 악하선 절제술 후 발생한 역설적 아랫 입술 경련

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## Paradoxical Lower Lip Twitching after Removal of the Submandibular Gland

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### = Abstract =

Benign submandibular gland tumor is a common disease in the head and neck region and can be completely removed by transcervical approach. Typical submandibular gland resection can lead to neurological complications. The most common complications are damage to the marginal mandibular branch of the facial nerve and weakening of the lower lip. We report a case of ipsilateral lower lip twitching occurring 3 months after surgery. There was no difficulty during the operation, and there were no neurologic complications immediate after surgery. However, in the present case, the patient experienced lower lip twitching paradoxically after surgery which has not been reported in the literature yet. Therefore, we reported this case with a review of relevant literature.

**Key Words :** Lip twitching · Submandibular gland

## Introduction

Benign tumors of the submandibular gland are common head and neck region tumors and they should be treated primarily by removal of the involved submandibular gland. Recurrence rate after surgery is very low. Generally, benign tumors of the submandibular gland are removed transcervically. This approach has a few complications, such as a neck scar and neurologic injury to the marginal mandibular, and hypoglossal nerves.<sup>1)</sup>

Lip twitching is caused by involuntary muscle contractions in or around the lips. These contractions can result from irritation of the affected nerves in the lips. Injuries to the nerves can also result in twitching and sometimes the nerves in the face do not function normally, causing the muscles in the lips to contract. When a lip twitches, the muscles in or around the area contract, which is evident to the patient as quivering of the lip.<sup>2,3)</sup> If the twitching occurs rapidly, the lip may not appear to be twitching to observers. In some cases, it may be an initial presentation of a degenerative condition of nerve. Muscle contraction in this region of the face can also be triggered by intense emotions of anger, fear, joy, and amusement, and can reflect extreme fatigue, stress, and anxiety.<sup>4-6)</sup>

We describe a case of a patient who presented with twitching of the lower lip while speaking and other facial expression changes after transcervical removal of the submandibular gland. We report this paradoxical movement of

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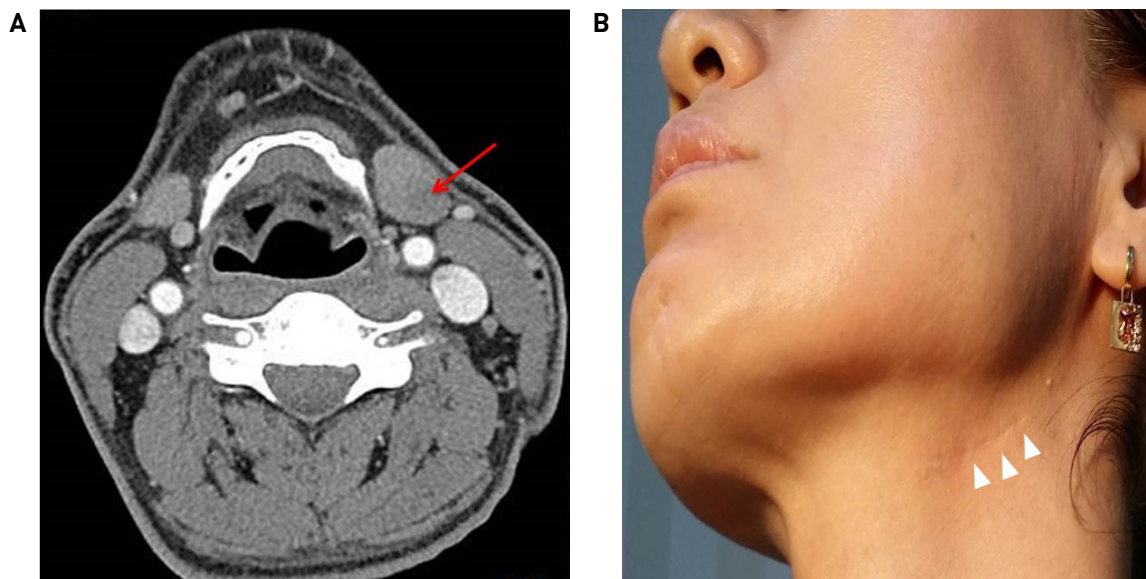
the lower lip with a review of relevant literature.

## Case Report

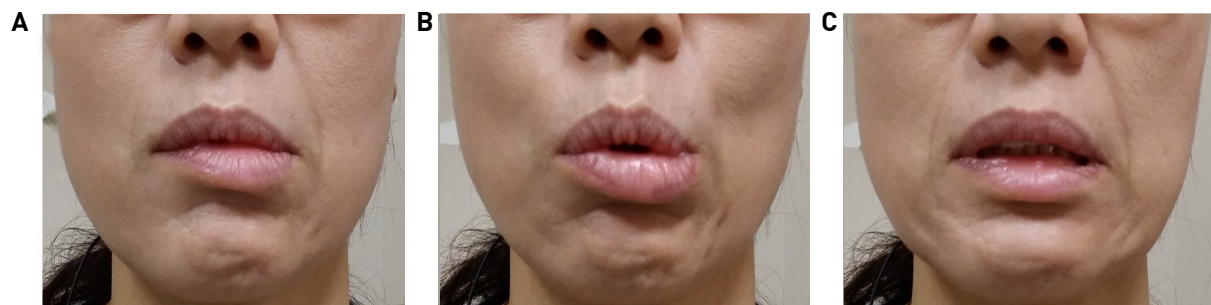
A 43-year-old female presented with a chief complaint of an enlarging left-submandibular mass during the previous 6 months. Her past history was not significant, with no history of surgery or use of tobacco or alcohol. Physical examination revealed a firm, mobile, and tender 3-4 cm size mass on the left submandibular area. No lesion was evident in the oral cavity, oropharynx, and other head and neck regions. The mandible, maxilla, and tooth were normal. A computed tomography (CT) scan with contrast showed a homogenous 3-4 cm size mass on level II of the left upper neck. The mass was evident as an intraglandular mass of the submandibular gland with no invasion to surrounding structures (Fig. 1). Radiologically, the mass was suspected of being

a benign tumor. The patient underwent needle aspiration (FNA) of the suspect benign pleomorphic tumor. The patient was scheduled for excision of submandibular gland. The mass was soft and well demarcated, with no severe adhesion to surrounding structures. We did not identify the marginal mandibular nerve to preserve the nerve intact. The submandibular gland was easily removed without any injury of the lingual and hypoglossal nerves. After surgery there were no complications, such as lower lip weakness, and lingual and hypoglossal nerve injury. Histologically, the benign mass was confirmed as pleomorphic adenoma.

Three months after surgery, the patient complained of left lower lip twitching when speaking and a changed facial expression (Fig. 2). There were neither palpable mass in the neck nor paralysis of the facial nerve suggesting no recurrence. The patient was observed for 6 months after surgery. The lower lip twitching did not improve. We rec-



**Fig. 1.** Computed tomography of the neck (A) shows homogenous hypodense mass (red arrow) in the submandibular gland, and scar (white arrow) on the left submandibular area (B).



**Fig. 2.** The photography of 3 months after the surgery. Mild lower lip deviation at resting (A), typical lip twitching at whistling (B) and lip widening (C).

ommended a Botox injection on the left lower lip. The patient denied this option and has been followed-up without any management.

## Discussion

Benign tumors of the submandibular gland are usually identified as painless masses and can be present for many years. Surgical excision is generally accepted as the management treatment of choice. Removal of a submandibular mixed tumor is commonly done using a transcervical approach. The procedure is relatively simple. The submandibular gland tissue can be removed without difficulty and without alteration of the salivary system. But, neurological complications can occur; injury of the mandibular branch of the facial nerve is common complication in the transcervical approach. On study reported an 18% incidence of damage to the marginal mandibular nerve, with damage being permanent in 7% of cases.<sup>1)</sup> The damage usually involves temporary paralysis due to a compression and /or stretching injury, which typically resolves spontaneously within 3 months.<sup>1)</sup>

Normally muscles contract when stimulated by the nerves and relax once this stimulation ceases. The strength of the contraction depends on the strength of nerve signals. However, in twitching the muscle contractions are sudden and rapid, imparting the sensation and/or appearance of lip trembling.<sup>3)</sup> Lip twitching is caused by involuntary, sudden muscle contractions in or around the lips. The visual symptoms of lower lip twitching can be very evident. A slight tremor can radiate to the involved lip following the muscle contractions. The main reasons are irritation of the facial nerve due to trauma or injury, and stimulation of the nerve endings that is present in the muscles. Injuries to the nerves can result in twitching; sometimes the nerves in the face function abnormally, causing the muscles in the lips to contract. When a lip twitches, the muscles in or around the area contract, making the lip feel like it is quivering.<sup>4)</sup> If the twitching is fast, the lip may not appear to be twitching to observers, but it may still be felt. Some conditions make the contractions more noticeable to others and may be accompanied by other symptoms. Other symptoms are mild palpitation-like movements, a feeling of frozen lips or numbness, and a curling sensation of the edges of lower lip. Twitching can occur repeatedly after an interval of several minutes.

Other suspected reasons are withdrawal from alcohol, drugs, or cigarettes, and venting of emotions like anger, excitement, fear, anxiety, or nervousness.

Delayed onset facial nerve complication, such as Bell's palsy or after parotidectomy, are major causes of abnormal facial movements. Bell's palsy is a transient condition of irritation or damage to facial nerve. Most people with Bell's palsy develop difficulty in talking, closing eye lids, and pursing lips due to muscular weakness. After recovery of long-term injury of facial nerve, synkinesis of facial nerves is possible. Synkinesis is the development of linked or unwanted facial movements.<sup>7)</sup> It is common and occurs in the majority of people who are recovering from prolonged facial palsy. Only the affected side of the face develops synkinesis. Synkinesis may be apparent in various ways. The recovering facial nerve is poorly insulated, resulting in messages to move one particular muscle being picked up by another muscle. The effect of this is more than one muscle moving at the same time (for example, an eye closing when smiling).<sup>8)</sup> Over time these linked movements become habitual and remain despite nerve recovery. Sometimes other muscles pull against the right muscles in a 'tug of war', which can result in a lack of movement. Synkinesis also often results in twitching movement, associated with aberrant regeneration following facial nerve injury.<sup>9)</sup> The most commonly used treatment for involuntary muscle contractions is local injections of botulinum toxin. Botulinum toxin inhibits the release of the neurotransmitter acetylcholine from axon endings at the neuromuscular junction resulting in local chemical denervation and loss of neuronal activity in the targeted organ. This markedly alleviates twitching movement, even though the abnormal muscle contraction does not completely disappear and electrophysiological signs of hyperexcitability remain.<sup>10)</sup>

## Conclusion

Lower lip weakness can occur in patients with excision of submandibular gland transcervically. However, in the present case, the patient experienced lower lip twitching paradoxically after surgery.

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